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Outcome Measures for Pediatric Laryngotracheal Reconstruction: International Consensus Statement

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Objectives: Develop multidisciplinary and international consensus on patient, disease, procedural, and perioperative factors, as well as key outcome measures and complications, to be reported for pediatric airway reconstruction studies.

Methods: Standard Delphi methods were applied. Participants proposed items in three categories: 1) patient/disease characteristics, 2) procedural/intraoperative/perioperative factors, and 3) outcome measures and complications. Both general and anatomic site-specific measures were elicited. Participants also suggested specific operations to be encompassed by this project. We then used iterative ranking and review to develop consensus lists via a priori Delphi consensus criteria.

Results: Thirty-three pediatric airway experts from eight countries in North and South America, Europe, and Australia participated, representing otolaryngology (including International Pediatric Otolaryngology Group members), pulmonology, general surgery, and cardiothoracic surgery. Consensus led to inclusion of 19 operations comprising open expansion, resection, and slide procedures of the larynx, trachea, and bronchi as well as three endoscopic procedures. Consensus was achieved on multiple patient/comorbidity (10), disease/stenosis (7), perioperative-/intraoperative-/procedure-related (16) factors.

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Consensus was reached on multiple outcome and complication measures, both general and site-specific (8 general, 13 supraglottic, 15 glottic, 17 subglottic, 8 cervical tracheal, 12 thoracic tracheal). The group was able to clarify how each outcome should be measured, with specific instruments defined where applicable.

Conclusion: This consensus statement provides a framework to communicate results consistently and reproducibly, facilitating meta-analyses, quality improvement, transfer of information, and surgeon self-assessment. It also clarifies expert opinion on which patient, disease, procedural, and outcome measures may be important to consider in any pediatric airway reconstruction patient.

Key Words: Airway reconstruction, pediatric, larynx, trachea, stenosis, Delphi, consensus.

Level of Evidence: 5.

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INTRODUCTION

Advances in open airway reconstructive techniques over the last four decades have improved our ability to avoid tracheostomy or decannulate patients with laryngeal and tracheal stenosis.[1–5] The dissemination of surgical techniques and medical protocols have allowed for these operations to be successfully performed worldwide rather than only at a few specialized centers. As a result, the number of open airway reconstructive procedures performed around the world appears to be increasing. Also, over this time, patients and the diseases with which they present have evolved in complexity, leading to a better understanding of surgical failure and the nuances of revision airway reconstruction.[6,7] Patients who have undergone multiple procedures or who have failed advanced endoscopic techniques present an ever-increasing level of complexity. Children who may have once been perceived as inoperable are now considered acceptable surgical candidates at many institutions.[7–9]

With more surgeons performing open airway reconstruction worldwide and the rising complexity of disease treated at many airway centers, the need for homogeneous outcome measures is of ever-increasing importance. In part, this need is due to the fact that historical measures of success, based primarily on the ability to maintain a patent airway or to decannulate a patient following surgery, have shifted over time. The outcomes of many investigations now span decades rather than focusing on early childhood or the postoperative period alone, thus highlighting the changes that occur to our patients throughout their lifetime.[10] In addition, post-reconstruction voice, swallow function, activity tolerance, and sleep patterns have become common topics of investigation and increasingly important to patients, adding intricacy and breadth to our interpretation of surgical outcomes.[4,11]

Despite an improved understanding of complex airway reconstruction and the long-term results of our interventions, outcome measures following laryngeal and tracheal surgery have become progressively more disparate. In an era of rapid data collection and transfer, and as our ability to share results among institutions and across continents in real time expands, this lack of uniform outcome measures has limited progress in the arena of open airway reconstruction. Because most institutions do not perform enough of these operations to independently analyze factors affecting outcomes, multicenter data collection or pooling of data are essential. However, these efforts also cannot move forward without agreement on key outcome measures and covariates to include in data collection.

The objective of this study is to define consensus outcome measures, including those pertaining to the patient, operation, disease, and associated complications for pediatric airway reconstructive surgery. Using the Delphi method, we sought to integrate the experience of an international multidisciplinary panel of airway surgeons to generate future hypotheses and improve communication of results across institutions. We also aim to generate a list of measures that any clinician caring for pediatric airway patients might consider when prospectively assessing outcomes of a reconstructive procedure. Because the Delphi method is iterative and results evolve during the study, we did not start with any specific hypotheses.

MATERIALS AND METHODS

In the original descriptions of the Delphi consensus method from the RAND Corporation in the 1960s, experts or stakeholders in a field completed surveys; their anonymized responses were pooled; and statistical summaries of these responses were provided as feedback to the participants.[12–14] Repeated cycles of this process led to gradual narrowing of response distributions, eventually resulting in consensus.[15] The methodology has been successfully adapted for use in healthcare,[16–19] although healthcare-related studies have often used a modified Delphi method that involves direct interaction and discussion among participating experts rather than pure statistical feedback, as originally described by RAND. Because data directed at determining the optimal choice of either general or site-specific variables and outcome measures are so limited, we chose to apply the Delphi method. However, because our experts were distributed across multiple continents and time zones, we elected to use the original Delphi method without direct interexpert discussion and interaction in order to allow efficient data collection and consensus development via email.

Forty pediatric airway specialists were recruited to this project, representing pediatric otolaryngology, pediatric pulmonology, general pediatric surgery, and pediatric cardiovascular surgery. All participants were selected for their established interest in pediatric airway reconstruction based on their clinical and scientific activities. No panelists had any relevant conflicts of interest.

Expert panelists were first asked to propose up to 20 items of focus in each of the following categories: 1) open laryngeal, tracheal, and bronchial operations for pediatric airway stenosis that should be covered by this consensus statement; 2) operation-specific and disease-specific considerations; and 3) patient-related and comorbidity-related considerations. Participants were asked to consider these items from the perspective of what should be reported in studies of airway reconstruction. A list for

each category was compiled and sent to all panelists for review in a series of four subsequent surveys. Each panelist rated each item on a 1 to 9 scale (9 = most important to include); and the median, mode, maximum, and minimum ratings for each item were calculated. Because the expert panel had a majority of otolaryngologists, we also calculated these data for that subset of panelists. Based on a previous Delphi consensus statement in otolaryngology,[20] we established a priori criteria for consensus (mean rating ≥ 7, with ≤ 1 response ≥ 2 points away from mean) and near consensus (mean rating ≥ 6.5, with ≤ 2 responses ≥ 2 points away from mean). These summary data were then sent out to the panelists, who then reviewed and re-ranked items with near-consensus status and those with mean scores above 6.5 but not reaching either consensus or near-consensus status. The results of the second round of ratings were considered final for each item category. All items not meeting consensus or near consensus were eliminated.

We then asked each panelist to propose up to 20 outcome measures and complications pertinent to the operations and procedures covered by this project. Items were proposed under one general category and five site-specific categories (supraglottis, glottis, subglottis, cervical trachea, thoracic trachea). The lists

TABLE I.
Airway Reconstruction Procedures Included in the Consensus Statement.

Procedure Category	Procedure Name	Reached Consensus for Inclusion?
Expansion operations	Single-stage laryngotracheoplasty	Yes
	Double-stage laryngotracheoplasty	Yes
	Open cricoid split	Yes
	Patch tracheoplasty	Yes
Resection operations	Single-stage tracheal resection	Yes
	Double-stage tracheal resection	Yes
	Single-stage partial cricotracheal resection	Yes
	Double-stage partial cricotracheal resection	Yes
	Single-stage extended cricotracheal resection	Yes
	Double-stage extended cricotracheal resection	Yes
	Bronchial sleeve resection	No
Slide operations	Cervical slide tracheoplasty	Yes
	Thoracic slide tracheoplasty	Yes
	Slide bronchoplasty	Yes
Glottic/supraglottic operations	Vocal cord lateralization	Yes
	Laryngeal web repair	Yes
	Open arytenoidectomy	Yes
	Open epiglottic petiole resuspension	Yes
Other operations	Open tracheal stent placement	Yes
	Open t-tube placement	Yes
	Tracheal homograft	Yes
	Tracheotomy	No
Operations for collapse or compression	External tracheal stent/splint	No
	External bronchial stent/splint	No
	Aortopexy	No
Voice/swallow operations	Tracheoesophageal fistula repair	No
	Laryngeal reinnervation	No
	Posterior cricoid reduction	No
Endoscopic operations	Balloon dilation	Yes
	Endoscopic cricoid split	Yes
	Endoscopic posterior graft laryngotracheoplasty	Yes
	Balloon dilation plus radial incisions	No
	Endoscopic tracheoesophageal fistula repair	No
	Supraglottoplasty	No
	Treatment for respiratory papillomatosis	No
	Cordotomy	No
Cyst/lesion excision operations	Saccular cyst excision	No
	Laryngocele excision	No
	Hemangioma excision	No

generated from this survey were sent to the panelists, who again rated each item 1 through 9. Summary data were calculated as above, and the lists and summary data then underwent a second round of ratings. The same consensus criteria were applied, and the consensus list of outcome measures and complications were sent to all panelists. Each panelist was asked to determine whether each item should be measured as a binary (yes/no), categorical, or continuous variable—and to suggest diagnostic studies, instrumental evaluations, or validated instruments relevant to that measure. Given the wide variety of responses regarding possible studies and instruments, we did not attempt to reach consensus for this segment of the study but simply identified common themes.

We did not discard the highest and lowest rating for each item in each survey because consensus and near-consensus criteria depended in part on assessing the number of outliers. Statistical analysis was performed using Excel 2010 (Microsoft Corp., Redmond, WA).

RESULTS

The project was conducted between June 2016 and October 2017. Of the 40 individuals invited to this project, 33 participated, representing 23 institutions in nine countries in North America, South America, Europe, and Australia. The 33 participants included three pediatric pulmonologists, 26 pediatric otolaryngologists, one pediatric cardiovascular surgeon, and three pediatric general surgeons. Invitees who expressed interest but did not return responses to the surveys included three pediatric cardiovascular surgeons and four pediatric otolaryngologists. With each survey, we used the same a priori criteria for consensus and near consensus.

Operations

The full list of operations proposed and those ultimately included are detailed in Table I. Twenty open operations of the larynx, trachea, and bronchi reached consensus for inclusion by the group. The panelists also reached consensus on inclusion of three endoscopic operations: balloon dilation, endoscopic cricoid split, and endoscopic posterior graft laryngotracheoplasty. Eight operations proposed were eliminated for not being targeted to treat airway stenosis, whereas bronchial sleeve resection was thought to be rare enough in children that the panelists chose not to include it. Five endoscopic operations were excluded by the group, as were three excision procedures for cysts and other lesions. Tracheotomy was excluded because it is not a reconstructive operation.

TABLE II.
Intraoperative and Perioperative Details of Reconstructive Operations.

Category	Item	Consensus Status
Patch/graft	Graft used: yes/no	Consensus
	Graft location: anterior, posterior, both, box graft	Consensus
	Graft type/source	Consensus
Stent/keel/endotracheal tube	Stent: yes/no	Consensus
	Stent duration	Consensus
	Stent type	Consensus
	Endotracheal tube duration (single-stage operation)	Consensus
	Keel: yes/no	Eliminated
	Keel duration	Eliminated
	Keel type	Consensus
Medication	Antibiotic use: therapeutic	Consensus
	Antibiotic use: prophylactic	Eliminated
	Perioperative steroid use/timing	Eliminated
Surgical approach	Single or double stage	Consensus
	Airway levels repaired	Consensus
	Use of cardiopulmonary bypass or ECMO	Consensus
	Surgical incision or approach	Eliminated
	Suturing: running vs. interrupted	Eliminated
Other operative details	Initial vs. revision surgery	Consensus
	Adjunct procedures required (e.g., balloon dilation)	Consensus
	Indication for surgery	Consensus
	Length of stay	Consensus
	Duration of follow-up	Near consensus
	Date of surgery	Eliminated

ECMO = extracorporeal membrane oxygenation.

TABLE III.
Airway, Comorbidity, and Patient-Related Factors Reaching Consensus or Near Consensus.

Airway Factors			Consensus Status
	Anatomic	Airway sizing	Consensus
		Distance from vocal folds to stenosis	Consensus
		Severity of stenosis (Myer-Cotton scale)	Consensus
		Length of stenosis	Consensus
		Location of stenosis (Monnier classification)	Near consensus
		Suprastomal collapse (yes/no)	Near consensus
	Functional	Vocal fold immobility: neurologic	Consensus
		Cricoarytenoid joint status	Consensus
		Static vs dynamic obstruction	Consensus
		Vocal fold immobility: cicatricial	Near consensus
	Other	Congenital vs acquired vs combined stenosis	Near consensus
Airway comorbidities	Sleep-disordered breathing	Tongue base obstruction	Near consensus
		Sleep apnea	Near consensus
		CPAP dependence	Near consensus
	Inflammation	Active/reactive larynx	Near consensus
		Mucosal inflammation and edema	Near consensus
	Central airway and bronchi	Airway compression	Consensus
		Tracheomalacia presence and severity	Consensus
		Bronchomalacia presence and severity	Consensus
	Prior airway history	Secondary airway lesions	Consensus
		Tracheostomy	Near consensus
		TEF history	Near consensus
		Indication for tracheostomy (airway vs neurologic vs pulmonary)	Near consensus
Other patient factors	General	Nutrition status	Near consensus
		Multidisciplinary team management (yes/no)	Near consensus
	Pulmonary/cardiac/thoracic	Aspiration status	Consensus
		Need for mechanical ventilation immediately prior	Consensus
		Bronchiectasis	Near consensus
		Pulmonary hypertension	Near consensus
		Chronic lung disease	Near consensus
		Cardiac disease	Near consensus
	GI/renal/hepatic	Documented GERD	Consensus
		Eosinophilic esophagitis	Near consensus
		Esophageal stricture	Near consensus
		Oral feeding status	Near consensus
	Syndromes/sequences	CHARGE	Consensus
		22q11/VCFS	Consensus
		Pierre Robin	Consensus
	Systemic	Wegener's (granulomatosis with polyangiitis)	Near consensus

CPAP = continuous positive airway pressure; GERD = gastroesophageal reflux disease; GI = gastrointestinal; TEF = tracheoesophageal fistula; VCFS = velocardiofacial syndrome.

Details of Reconstructive Operations

Details of the operations can be seen in Table II. A total of 16 items reached consensus, and one item reached near consensus. These items were divided into patch-/graft-related, stent-/keel-/endotracheal tube-related, medication-related, surgical approach-related, and other operation-related categories. Another seven items failed to reach consensus or near-consensus status and were eliminated.

Airway-, Disease-, and Patient-Related Factors

Seven airway items reached consensus and four near consensus, as demonstrated in Table III. These items were categorized as anatomic, function-related, and other. Under the domain of airway-related comorbidities, 17 items were proposed, of which four reached consensus and eight reached near consensus. These were further categorized as sleep apnea-related, voice-related, inflammation-related, central airway and bronchi-related, and prior procedure-/

TABLE IV.
General Outcome Measures.

Measure Name	Consensus Status	How to Measure
Mortality	Consensus	Categorical: yes/no and airway-related/ unrelated
Need for revision	Consensus	Binary: yes/no
Number of subsequent open procedures	Consensus	Continuous
Number of subsequent endoscopic procedures	Consensus	Continuous
Number of dilations	Consensus	Continuous
Need for adjunctive procedures	Consensus	Binary: yes/no
Long-term patency (10-year)	Consensus	Binary: yes/no
Quality of life	Consensus	General vs disease-specific measures
Parent/caregiver satisfaction	Near consensus	Future PROMs/PREMs
Patient satisfaction	Near consensus	Future PROMs/PREMs
Duration of follow up	Eliminated	
ICU days	Eliminated	
Hospital days	Eliminated	
Wound infection	Eliminated	
Duration of mechanical ventilation	Eliminated	
Duration of sedation	Eliminated	
Sedation withdrawal	Eliminated	
Weight gain	Eliminated	

ICU = intensive care unit; PROM = patient-reported outcome measure; PREM = patient-reported experience measure.

injury-/or disease-related. Panelists also considered 27 other patient factors, with six reaching consensus and 10 near consensus, in the categories of general, pulmonary/ cardiopulmonary/thoracic, GI/renal/hepatic, syndrome-/ sequence-related, and systemic/infectious.

Outcome Measures

The panelists proposed 172 outcome measures and complications, both general and site-specific. Eight general measures led to consensus for eight and near consensus for two (Table IV).

Site-specific measures were divided into complications/adverse events, surgical site outcomes, respiratory outcomes, voice outcomes, and swallow/feeding outcome. For the supraglottis, 25 site-specific measures were proposed (13 consensus, 2 near consensus). Thirty-one glottic measures were proposed (15 consensus, 2 near consensus). Thirty-seven proposed subglottic measures led to 17 consensus and six to near consensus. Consensus and near-consensus outcome measures for these three laryngeal sites are presented in Tables V and VI.

Finally, 29 cervical tracheal measures led to eight consensus and four near consensus, whereas 32 thoracic tracheal items led to 12 consensus and seven to near consensus (Tables VII and VIII).

The panelists' views were quite varied on how best to define and measure each item in the general and site-specific outcome lists. As such, we did not attempt to reach consensus for this issue but instead attempted to identify trends. Panelists were able to identify validated instruments for some items. However, many items did not have such an instrument extant, leading to the proposal for either imaging or instrumental evaluations for many measures. These are demonstrated in Tables IV through VIII for each item.

DISCUSSION

Published case numbers for open pediatric airway reconstruction worldwide are lacking; however, experience and anecdotal data suggest that the incidence of these procedures has been increasing for several reasons. Foremost, the survival of premature infants and critically ill children requiring prolonged intubation or tracheostomy has increased, leading to a greater frequency of complex, and often revision, reconstructive procedures. In addition, with the dissemination of experienced surgeons worldwide, more institutions are performing open pediatric airway reconstruction rather than tracheostomy. Despite this increase, the majority of single institutions do not perform enough of these operations annually to allow effective single-center analysis of factors affecting outcomes and complications. As a result, multicenter collection of prospective data and pooled analysis of retrospective data are essential. These efforts, however, cannot move forward without standardization of outcome measures, disease and comorbidity measures, and reporting of complications. Equally important are standardized descriptors of the reconstructive procedures themselves. This study aims to address these needs in the absence of data to direct the selection of these measures, using a structured expert consensus Delphi approach. It is the first study to generate international consensus on which measures and data are the most important when reporting outcomes of open pediatric airway reconstruction. We

TABLE V.
Site-Specific Outcome Measures for Laryngeal Reconstruction, Including the Supraglottis and Glottis.

Anatomic Site	Category	Measure Name	Consensus Status	How to Measure
Supraglottis	Complications and adverse events	Need for unplanned tracheostomy replacement or delayed tracheostomy	Consensus	Binary: yes/no
		Need for unplanned stent replacement or delayed stent placement	Consensus	Binary: yes/no
	Surgical site	Airway size	Consensus	Continuous
		Cricoarytenoid joint fixation	Consensus	Direct laryngoscopy with palpation, flexible laryngoscopy Binary
		Posterior prolapse of epiglottis/petiole prolapse	Consensus	Flexible endoscopy, sleep-state endoscopy Categorical: none/mild/moderate/severe
		Scarring	Eliminated	
		Presence/absence of anterior glottic webbing	Eliminated	
		Postoperative granulation tissue	Eliminated	
		False fold shortening	Eliminated	
	Respiratory	Tracheostomy decannulation	Consensus	Binary
		Obstructive sleep apnea	Consensus	PSG Categorical: none/mild/moderate/severe
		Need for noninvasive respiratory support	Consensus	Binary: yes/no
		Exercise tolerance	Consensus	Exercise laryngoscopy, subjective rating Categorical: full-exercise tolerance; limited with extreme exertion; limited with any exertion; limited at rest
		Apnea–hypopnea index	Near consensus	PSG Continuous
		Time to decannulation	Eliminated	
		Sleep disturbance	Eliminated	
		Cough	Eliminated	
	Voice	Presence/absence of supraglottic voicing	Consensus	Binary
		Clinical voice evaluation	Near consensus	PVHI, CAPE-V, stroboscopy
		Voice quality	Eliminated	
		Voice analysis	Eliminated	
		Phonation time	Eliminated	
	Swallow/feeding	Aspiration	Consensus	VFSS, FEES Categorical: none/mild/moderate/severe
		Ability to eat orally	Consensus	Categorical: limitations and dietary modifications
		Dysphagia	Consensus	Binary vs. categorical
Glottis	Complications and adverse events	Need for unplanned tracheostomy replacement or delayed tracheostomy	Consensus	Binary: yes/no
		Need for unplanned stent replacement or delayed stent placement	Consensus	Binary: yes/no
	Surgical site	Airway size	Consensus	Continuous
		Cricoarytenoid joint fixation	Consensus	Direct laryngoscopy with palpation, flexible laryngoscopy Binary
		Graft-related complications	Consensus	Binary Graft loss, prolapse
		Arytenoid prolapse	Consensus	Flexible laryngoscopy, exercise laryngoscopy, sleep-state endoscopy Categorical: static/dynamic; none/mild/moderate/severe
		% webbing	Near consensus	Continuous
		Vocal fold height mismatch	Eliminated	
		Presence/absence of anterior glottic webbing	Eliminated	
		Postoperative granulation tissue	Eliminated	
		V-shaped anterior commissure	Eliminated	
	Respiratory	Tracheostomy decannulation	Consensus	Binary: yes/no
		Adequacy of airway	Consensus	
		Retractions	Consensus	Categorical: none/mild/moderate/severe; at rest/sleeping/with activity
		Stridor	Consensus	

(Continues)

TABLE V.
(Continued)

Anatomic Site	Category	Measure Name	Consensus Status	How to Measure
				Categorical: none/mild/moderate/severe; at rest/sleeping/with activity
		Need for noninvasive respiratory support	Consensus	Binary
		Exercise tolerance	Consensus	Exercise laryngoscopy, subjective rating Categorical: full-exercise tolerance; limited with extreme exertion; limited with any exertion; limited at rest
		Time to decannulation	Eliminated	
		Apnea–hypopnea index	Eliminated	
		Cough	Eliminated	
	Voice	Vocal fold mobility	Consensus	Flexible laryngoscopy, stroboscopy Categorical: normal/hypomobile/immobile; unilateral/bilateral
		Clinical voice evaluation	Near consensus	PVHI, CAPE-V, stroboscopy
		Voice quality	Eliminated	
		Presence/absence of supraglottic voicing	Eliminated	
		Voice analysis	Eliminated	
		Mucosal wave	Eliminated	
	Swallow/feeding	Ability to eat orally	Consensus	Categorical: limitations and dietary modifications
		Aspiration	Consensus	VFSS, FEES Categorical: none/mild/moderate/severe
		Dysphagia	Eliminated	
		Removal of gastrostomy tube	Eliminated	
		Dietary restrictions	Eliminated	

CAPE-V = Consensus Auditory-Perceptual Evaluation of Voice; FEES = functional endoscopic evaluation of swallow; PSG = polysomnography; PVHI = Pediatric Voice Handicap Index; VFSS = videofluoroscopic swallow study.

propose that these measures may also be useful when counseling patients and assessing results for any single individual undergoing such an operation or any single surgeon's practice. We aimed to include both generalized measures that are applicable to all patients undergoing airway reconstruction as well as specific measures that may differ between patients based on surgical site.

One aim of this study was to create a consensus list of manageable length such that it might be realistically used in research and clinical practice. By dividing items into general and site-specific categories, we were able to achieve this goal while still addressing multiple key areas such as surgical site outcomes, respiratory/airway status, voice, and swallow. Similarly, the list of items addressing patient and disease factors was reduced to 17 consensus measures, which we again think is practically usable. An operation that addresses multiple sites in the airway will of course require a longer list of measures, although the overlap across anatomic sites will somewhat mitigate this issue.

This study has several strengths. The panel of participants includes multiple international and national experts from four major specialties involved in pediatric airway reconstruction. These experts represent multiple countries and institutions around the world. Participants also represent a wide range of career stages, from relatively junior clinicians to veteran clinicians with decades of experiences. The number of participants is also very large for a Delphi study. By incorporating diverse perspectives, we have produced a consensus that will hopefully be both acceptable and credible to the wide range of providers involved in planning and executing these operations. This broad range of expertise notwithstanding, we found more agreement across participants and specialties than we had expected. Despite the number of items proposed and evaluated in each stage of this study, we also found that survey fatigue was not a major problem. We found that the same items reached different results for different anatomic sites, suggesting that participants did not literally or figuratively copy and paste ratings from section to section. Methodologically, the use of a priori definitions of consensus and near consensus allowed clear determination of these results. Meanwhile, the intentional lack of interparticipant discussion allowed each voice to count equally, rather than allowing more experienced or well-known individuals to disproportionately influence others' ratings. Finally, we found that several items initially expected to reach consensus did not. For example, methicillin-resistant Staphylococcus aureus and Pseudomonas colonization status and Trisomy 21 were eliminated as patient-related factors of interest to the group, whereas tracheostomy reached only near consensus. Although such findings require further evaluation, we suggest that they are important because they bring into question conventional wisdom and practice, which in turn may lead to improvement in clinical management associated with airway reconstructive operations.

TABLE VI.
Site-Specific Outcome Measures for Subglottic Laryngeal Reconstruction.

Category	Measure Name	Consensus Status	How to Measure
Complications and adverse events	Need for unplanned tracheostomy replacement or delayed tracheostomy	Consensus	Binary: yes/no
	Need for unplanned stent replacement or delayed stent placement	Consensus	Binary: yes/no
Surgical site	Restenosis/failure	Consensus	Binary: yes/no
	Airway size	Consensus	Categorical (Myer-Cotton scale)
	Loss of graft	Consensus	Binary: yes/no
	Dynamic collapse/A-frame deformity	Consensus	Flexible bronchoscopy, exercise laryngoscopy Binary vs. categorical: none/mild/moderate/severe
	Change in airway sizing	Consensus	Categorical (Myer-Cotton scale)
	Graft-related complications	Consensus	Binary Graft loss, prolapse
	Cricoarytenoid joint fixation	consensus	Direct laryngoscopy with palpation, flexible laryngoscopy Binary: yes/no
	Arytenoid prolapse	Consensus	Flexible laryngoscopy, exercise laryngoscopy, sleep state endoscopy Categorical: static/dynamic, none/mild/moderate/severe
	Airway growth over time	Consensus	Categorical (Myer-Cotton scale)
	Dehiscence	Near consensus	Binary: yes/no
	Postoperative granulation tissue	Eliminated	
Respiratory	Tracheostomy decannulation	Consensus	Binary: yes/no
	Stridor at rest	Consensus	Binary: yes/no
	Exercise tolerance	Consensus	Exercise laryngoscopy, subjective rating Categorical: full-exercise tolerance, limited with extreme exertion, limited with any exertion, limited at rest
	Need for noninvasive respiratory support	Consensus	Binary: yes/no
	Time to decannulation	Near consensus	Continuous
	Retractions	Near consensus	Categorical: none/mild/moderate/severe, at rest/sleeping/with activity
	Cyanotic or apneic episodes	Near consensus	Binary: yes/no
	Number of reintubations	Eliminated	
	Stridor on exertion	Eliminated	
	Apnea–hypopnea index	Eliminated	
	Episodes of croup	Eliminated	
	Obstructive sleep apnea	Eliminated	
	Pulmonary function testing/fixed obstruction on spirometry	Eliminated	
	Sleep disturbance	Eliminated	
	Exercise testing	Eliminated	
	Respiratory infections	Eliminated	
	Cough	Eliminated	
Voice	Vocal fold mobility	Consensus	Flexible laryngoscopy, stroboscopy Categorical: normal/hypomobile/immobile; unilateral/bilateral
	Clinical voice evaluation	Near consensus	PVHI, CAPE-V, stroboscopy
	Voice analysis	Eliminated	
	Stroboscopy	Eliminated	
Swallow/feeding	Aspiration	Consensus	VFSS, FEES Categorical: none/mild/moderate/severe
	Dysphagia	Near consensus	Binary vs. categorical
	Removal of gastrostomy tube	Eliminated	

CAPE-V = Consensus Auditory-Perceptual Evaluation of Voice; FEES = functional endoscopic evaluation of swallow; PVHI = Pediatric Voice Handicap Index; VFSS = videofluoroscopic swallow study.

Despite the aforementioned strengths of the study, several limitations merit mention. Foremost, the Delphi method itself is designed to generate expert consensus in the absence of clear foundational data. It is therefore, by definition, level 5 evidence and is intended to serve until more high-level data become available. We hope that this study provides the impetus for studies to generate those data and to validate the measures proposed here. The

TABLE VII.
Site-Specific Outcome Measures for Cervical Tracheal Reconstruction.

Category	Measure Name	Consensus Status	How to Measure
Complications and adverse events	Need for unplanned tracheostomy replacement or delayed tracheostomy	Consensus	Binary: yes/no
	Need for unplanned stent replacement or delayed stent placement	Consensus	Binary: yes/no
	Need for postoperative ECMO	Eliminated	
Surgical site	Restenosis/failure	Consensus	Binary: yes/no
	Airway size	Consensus	Continuous
	Loss of graft	Consensus	Binary: yes/no
	Proximal tracheomalacia	Near consensus	Flexible bronchoscopy, exercise laryngoscopy Binary vs. categorical: none/mild/moderate/severe
	Postoperative granulation tissue	Eliminated	
Respiratory	Exercise tolerance	Consensus	Exercise laryngoscopy, subjective rating Categorical: full-exercise tolerance, limited with extreme exertion, limited with any exertion, limited at rest
	Retractions	Near consensus	Categorical: none/mild/moderate/severe, at rest/sleeping/ with activity
	Need for noninvasive respiratory support	Near consensus	Binary: yes/no
	Stridor at rest	Near consensus	Binary: yes/no
	Wheezing/respiratory distress	Eliminated	
	Stridor on exertion	Eliminated	
	Apnea–hypopnea index	Eliminated	
	Time to decannulation	Eliminated	
	Cyanotic or apneic episodes	Eliminated	
	Recurrent bronchitis or pneumonia	Eliminated	
	Recurrent respiratory issues	Eliminated	
	Exercise testing	Eliminated	
	Pulmonary function testing/fixed obstruction on spirometry	Eliminated	
	Respiratory infections	Eliminated	
	Peak-flow measurements	Eliminated	
	Episodes of croup	Eliminated	
	Cough	Eliminated	
Voice	Vocal fold paralysis	Consensus	Flexible laryngoscopy, stroboscopy, EMG Categorical: normal/hypomobile/immobile, unilateral/bilateral
	Vocal fold mobility	Consensus	Flexible laryngoscopy, stroboscopy Categorical: normal/ hypomobile/immobile, unilateral/bilateral
	Clinical voice evaluation	Eliminated	
Swallow/feeding	Removal of gastrostomy tube	Eliminated	

ECMO = extracorporeal membrane oxygenation; EMG = electromyography.

expert panel did not include as many cardiovascular surgeons as were invited. We also did not have a practical method to determine how including further participants might alter the results of this study. Furthermore, the panel was clearly dominated by otolaryngologists. Whereas this majority might reflect the global distribution of providers performing these operations, it might also bias the results. We attempted to counter this by providing participants both overall and otolaryngology-specific summary results for each item rated; however, we have no way to determine whether this approach was necessary or whether it altered our results. Similarly, we were not able to include participants from African or Asian countries, despite invitations to experts from those continents. Our participants also uniformly represented resource-rich nations and institutions, which may have affected the results of our consensus, for example, by allowing more expensive studies and procedures to be included. We cannot easily measure the magnitude or direction of this bias. With regard to the results themselves, we did not attempt to reach structured consensus on how to measure any given outcome or complication. We would suggest that such consensus might be pursued in future studies.

We acknowledge that wide implementation of these reporting standards may take several years. Any study that is to be published in the near future will include patients recruited prior to publication of this consensus statement, and prospective collection of data using the measures outlined here will require gradual adoption

TABLE VIII.
Site-Specific Outcome Measures for Thoracic Tracheal Reconstruction.

Category	Measure Name	Consensus Status	How to Measure
Complications and adverse events	Need for unplanned tracheostomy replacement or delayed tracheostomy	Consensus	Binary: yes/no
	Need for unplanned stent replacement or delayed stent placement	Consensus	Binary: yes/no
	Need for postoperative ECMO	Consensus	Binary: yes/no
Surgical site	Restenosis/failure	Consensus	Binary: yes/no
	Dehiscence	Consensus	Binary: yes/no
	Airway size	Consensus	Continuous
	Fistula	Consensus	Binary: yes/no
	Recurrent laryngeal nerve damage	Consensus	EMG, flexible laryngoscopy Binary vs. categorical: normal/hypomobile/immobile, unilateral/bilateral
	Figure-8 deformity causing significant stenosis	Consensus	Bronchoscopy Binary: yes/no
	Extrinsic airway compression	Near consensus	Bronchoscopy, CT angiography Binary: yes/no
	Tracheomalacia	Near consensus	Flexible bronchoscopy, exercise laryngoscopy Binary vs categorical: none/mild/moderate/severe
	Airway growth over time	Near consensus	Continuous
	Tracheal growth measured by z-score relative to normal size for age	Near consensus	Continuous
	Postoperative granulation tissue	Eliminated	
	Phrenic nerve injury	Eliminated	
Respiratory	Need for noninvasive respiratory support	Consensus	Binary: yes/no
	Exercise tolerance	Consensus	Exercise laryngoscopy, subjective rating Categorical: Full-exercise tolerance, limited with extreme exertion, limited with any exertion, limited at rest
	Cyanotic or apneic episodes	Near consensus	Binary: yes/no
	Wheezing/respiratory distress	Near consensus	Categorical: none/mild/moderate/severe, need for airway clearance
	Time to decannulation	Near consensus	Continuous
	Recurrent bronchitis or pneumonia	Eliminated	
	Stridor	Eliminated	
	Apnea–hypopnea index	Eliminated	
	Pulmonary function testing/fixed obstruction on spirometry	Eliminated	
	Recurrent respiratory issues	Eliminated	
	Peak-flow measurements	Eliminated	
	Washing-machine breathing	Eliminated	
	Cough	Eliminated	
Voice	Vocal fold paralysis	Consensus	Flexible laryngoscopy, stroboscopy Categorical: normal/hypomobile/immobile, unilateral/bilateral
	Voice quality	Eliminated	
	Vocal fold mobility	Eliminated	
	Clinical voice evaluation	Eliminated	
Swallow/feeding	None		

CT = computed tomography; ECMO = extracorporeal membrane oxygenation; EMG = electromyography.

among pediatric airway specialists around the world. In the meantime, our next objectives will be to perform similar studies for pediatric voice and swallow operations. We are also working to develop the findings of the current study into an international database or registry. In beginning that process, we have encountered significant hurdles, including questions of data integrity, data quality assurance, who should own and control access to any data, national and international regulatory issues, and potential medicolegal issues such as discoverability. Furthermore, a critical question is how to balance any regulatory requirements and costs of operating such a registry with the need to facilitate access for surgeons in resource-limited settings.

In the meantime, we hope that this article will inspire researchers to develop and validate measures for the items in Tables IV through VIII that lack well-defined instruments or tools. We also invite interested surgeons from multiple specialties to contact us about participating in future iterations and updates of this project.

CONCLUSION

This consensus statement provides a framework for future studies to communicate results consistently and reproducibly. Key disease, patient, and comorbid factors were identified, as were important details of operative and perioperative management, outcomes, and complications. Both general and site-specific measures were included. This development will facilitate meta-analyses, quality improvement, transfer of information, and surgeon self-assessment. It also clarifies expert opinion on which patient, disease, procedural, and outcome measures may be important to consider in any pediatric airway reconstruction patient. Future work will clarify how best to measure each item included in this statement and will validate these approaches to measurement.

BIBLIOGRAPHY

1. Cotton RT, Myer CM 3rd, O'Connor DM. Innovations in pediatric laryngotracheal reconstruction. *J Pediatr Surg* 1992;27:196–200.
2. Cotton RT. Management of subglottic stenosis. *Otolaryngol Clin North Am* 2000;33:111–130.
3. Rutter MJ, Hartley BE, Cotton RT. Cricotracheal resection in children. *Arch Otolaryngol Head Neck Surg* 2001;127:289–292.
4. Jefferson ND, Cohen AP, Rutter MJ. Subglottic stenosis. *Semin Pediatr Surg* 2016;25:138–143.
5. Jacobs IN, Redden RA, Goldberg R, et al. Pediatric laryngotracheal reconstruction with tissue-engineered cartilage in a rabbit model. *Laryngoscope* 2016;126(suppl 1):S5–S21.
6. de Alarcon A, Rutter MJ. Revision pediatric laryngotracheal reconstruction. *Otolaryngol Clin North Am* 2008;41):959–980.
7. de Alarcon A, Rutter MJ. Cervical slide tracheoplasty. *Arch Otolaryngol Head Neck Surg* 2012;138:812–816.
8. Kopelovich JC, Wine TM, Rutter MJ, Mitchell MB, Prager JD. Secondary reverse slide tracheoplasty for airway rescue. *Ann Thorac Surg* 2016;101: 1205–1207.
9. Yamamoto K, Monnier P, Holtz F, Jaquet Y. Laryngotracheal reconstruction for pediatric glotto-subglottic stenosis. *Int J Pediatr Otorhinolaryngol* 2014;78:1476–1479.
10. Pullens B, Dulfer K, Buysse CM, Hoeve LJ, Timmerman MK, Joosten KF. Long-term quality of life in children after open airway surgery for laryngotracheal stenosis. *Int J Pediatr Otorhinolaryngol* 2016;84:88–93.
11. Zacharias SR, Weinrich B, Brehm SB, et al. Assessment of vibratory characteristics in children following airway reconstruction using flexible and rigid endoscopy and stroboscopy. *JAMA Otolaryngol Head Neck Surg* 2015;141:882–887.
12. Fink A, Kosecoff J, Chassin M. *Consensus Methods: Characteristics and Guidelines for Use*. Santa Monica, CA: RAND Corporation; 1991.
13. Dalkey NC. *The Delphi Method: An Experimental Study of Group Opinion*. Santa Monica, CA: RAND Corporation; 1969.
14. Brown BB. *Delphi Process: A Methodology Used for the Elicitation of Opinions of Experts*. Santa Monica, CA: RAND Corporation; 1968.
15. Cline A. *Prioritization Process Using Delphi Technique*. Dublin, OH: Carolla Development; 2000.
16. Ahmed A, Ishman SL, Laeeq K. Assessment of improvement of trainee surgical skills in the operating room for tonsillectomy. *Laryngoscope* 2013; 123:1639–1644.
17. Hill P, Normal G, Davison T. Adult bone anchored hearing aid services in the United Kingdom: building a consensus for development. *Cochlear Implants Int* 2012;13:228–236.
18. Mitchell RB, Hussey HM, Setzen G. et al. Clinical consensus statement: tracheostomy care. *Otolaryngol Head Neck Surg* 2013;148:6–20.
19. Balakrishnan K, Bauman N, Chun RH, et al. Standardized outcome and reporting measures in pediatric head and neck lymphatic malformations. *Otolaryngol Head Neck Surg* 2015;152:948–953.
20. Rhee JS, Weaver EM, Park SS, et al. Clinical consensus statement: diagnosis and management of nasal valve compromise. *Otolaryngol Head Neck Surg* 2010;143:48–59.